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PAEDIATRIC CAUSE OF BACK PAIN IN YOUNG ADULT. A CASE REPORT.

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INTRODUCTION

Eosinophilic granuloma is a non-neoplastic lesion commonly occurs in paediatric age group, arising from clonal proliferation of Langerhans-type histiocytes. Vertebral involvement has been frequently reported in approximately 10-15% of cases, but involvement of the spinal cord and roots remains a rare occurrence. Patients may present with progressive back pain, with or without neurologic symptoms. We describe an additional case of Langerhans cell histiocytosis (LCH) of the spine in a young adult presented with a progressive back pain with quadriparesis.

CASE REPORT

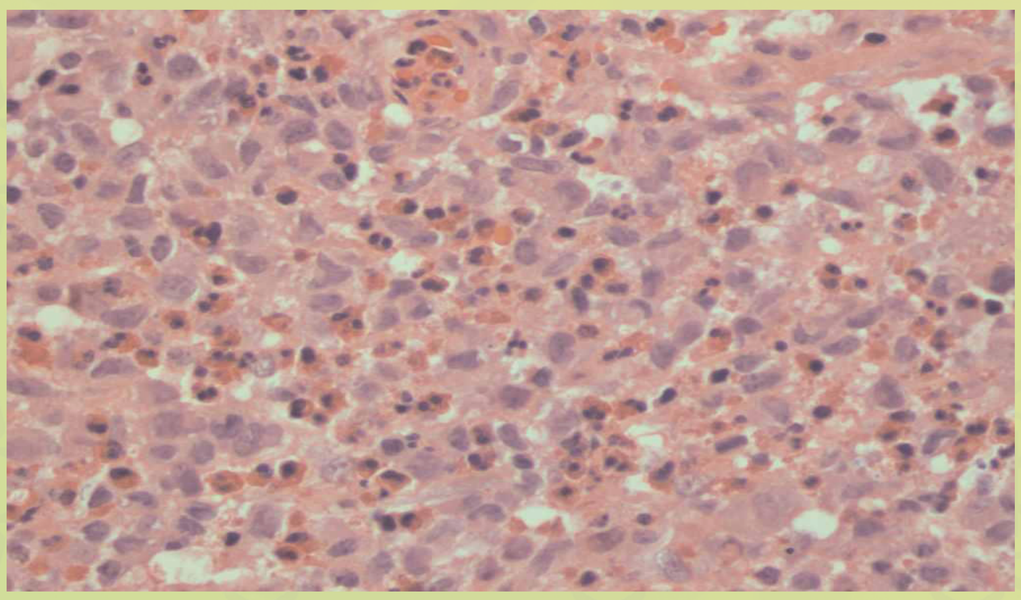
A 37 years old man presented with a 5-month history of progressive back pain associated with hemiparesis. Pain was constant, pricking in nature with increasing in intensity at night. He needed support to walk. He had no difficulty with control of sphincters. There was no other constitutional symptom. No history of spinal trauma and denied any contact history with tuberculosis. Family and medical history were unremarkable. On examination, the cranial nerves were intact. There was tenderness over the spinous process of the thoracic vertebrae. Sensation was reduced over the right side at the level of T7 and T8 with presence of clonus and hypereflexia. Voluntary power in all muscle groups was fairly normal. No lymphadenopathy was found. Other systemic reviews were unremarkable.

Chest x-ray and blood tests were in the normal range, except for a slight elevation of total white cell and ESR of 70mm/h. Mantoux test was positive. Plain radiograph showed disruption of pedicles T6/T7 with osteolytic changes in T4-T6, L3-L4, S1, and vertebral collapse in L4. MRI showed features suggestive of multiple levels of vertebrae and ilium destruction. In view of patient's age and available results, our initial impression was spinal tuberculosis, with metastasis and lymphoma among the differentials considered.

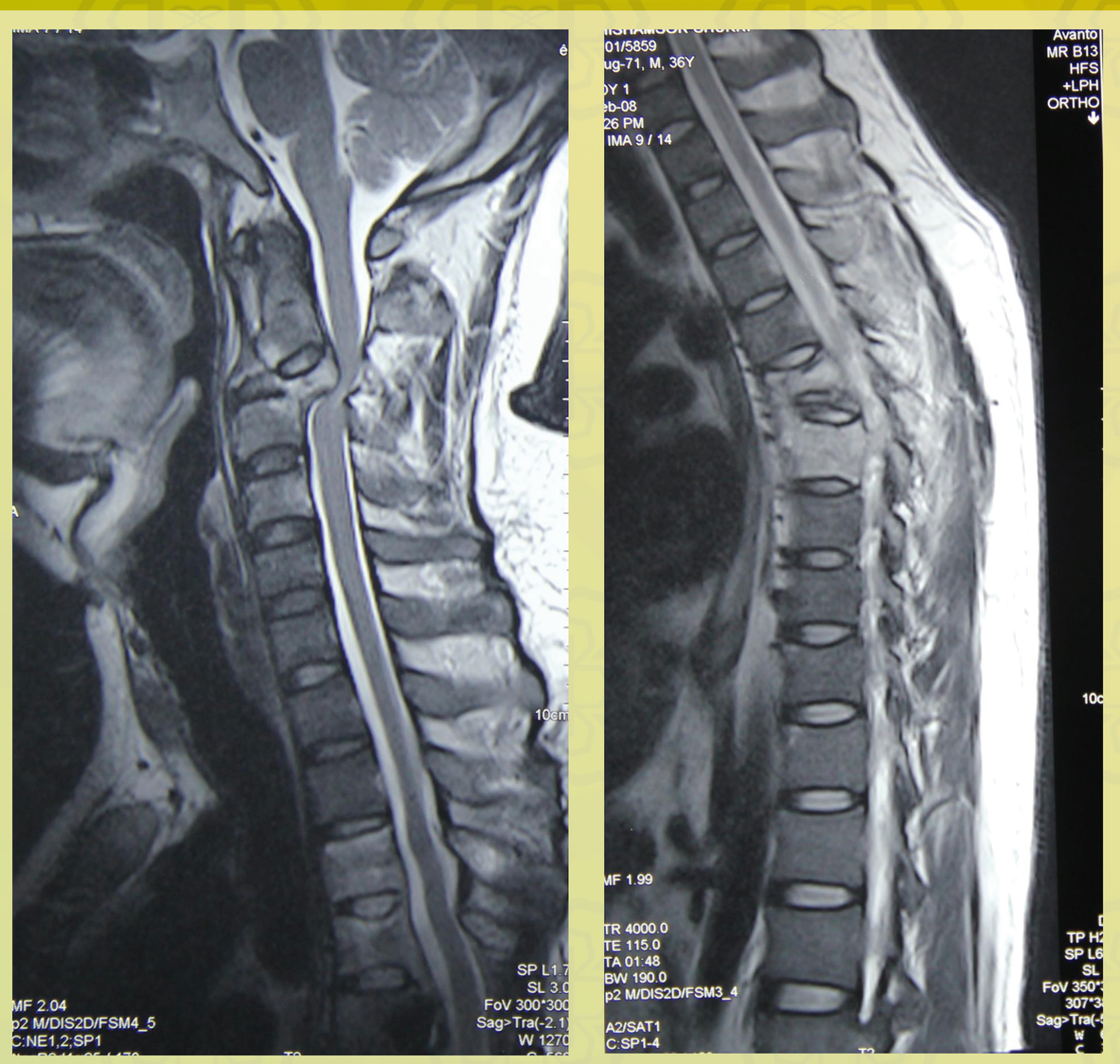
We performed a posterior decompression and posterior instrumentation with bone biopsy of the T6 and T7 vertebrae. Intra-operatively, there was presence of abnormal soft, gray-colored extrathecal tissue with destruction and fragile T6 and T7 vertebral bodies. HPE reported as chronic inflammation with no evidence of malignancy or infection. After surgery, he was started on anti-TB regime and responded well. On regular follow ups, patient was ambulating well and able to perform daily activities independently.

17 months post surgery, he presented again with quadriparesis and unable to micturate. On examination, power of both upper limb and lower limb was 3/5 with reduced sensation from C6 and below. Repeated MRI showed subluxation of C3/C4, destruction of C3 body and reduction of C4-C5 vertebral height. There was also cord compression secondary to the C3/C4 subluxation. Previous bone and tissue samples were re-evaluated and revealed morphological features and immunohistochemistry analysis consistent with LCH. He was then subjected for posterior decompression and instrumentation of C2-C4. New biopsy was taken and confirmed the diagnosis.

He was well after the second surgery. On subsequent follow-up, patient showed great improvement and back to his pre-disease daily activities.



Histological appearance revealed abundant of histiocytes



MRI of cervical and lumbar spine, showing collapse of vertebral height compressing the spinal cord.

DISCUSSION

LCH is regarded as a paediatric disease. However, recent studies have reported approximately 30% of cases in adults. In the adult population, LCH may present a diagnostic dilemma; there are great numbers of relatively commoner lesion with a similar appearance and a low index of suspicion for LCH among practitioners.

Although LCH of the spinal column is a frequent finding, very few cases of spinal cord or root involvement have been reported in the literature. From a clinical standpoint, patients affected by spinal LCH experience pain usually in the thoracic or the lumbar region. Neurological deficits occur only rarely.

From a radiological standpoint, almost invariably reveal pathological changes of the vertebral body only; The degree of involvement of the vertebral body may result in a vertebra plana. These features are frequently encountered in other spinal tumors, both primitive and metastatic, and differential diagnosis should be made on the basis of a biopsy and a search for the aforementioned specific histological and immunological features. Our patient exhibits a similar dilemma in making the correct diagnosis.

CONCLUSION

LCH is rare in adults but should not be ruled out. Difficult clinical presentation and histology requires higher degree of suspicion. This presentation is to emphasize the importance of including LCH in the differentials of radioluscent lesions of the bone in adults.

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